Annals of Clinical & Analytical Medicine
Review

Mandibular Buccal Bifurcation Cyst with Radiologic and Histopathologic Features: Two Case Reports and a Literature Review

Mandibular buccal bifurcation cyst features: Two case reports and literature review

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Abstract
This article aims to present the clinical, radiological, histological features and diagnostic criteria of mandibular buccal bifurcation cyst (MBBC) while adding two new cases to the literature. In our study, literature search was made on PubMed and Google Scholar. The following words were used for the search: (buccal bifurcation"OR"inflammatory collateral"OR paradental) and cyst. In addition, two cases were presented. The diagnosis of the cases was made by cone beam computed tomography (CBCT) and histological findings together. Histopathologic features of MBBC is identical to inflammatory odontogenic cysts. Therefore, the diagnosis should be made by the combination of clinical, radiographic, and histopathological features. Knowing this entity, especially in childhood, will guide clinicians in protecting the affected tooth while performing cyst treatment in most of the cases.

Keywords
Mandibular Buccal Bifurcation Cyst; Inflammatory Odontogenic Cyst; Inflammatory Collateral Cyst; Odontogenic Cyst; Cone Beam Computed Tomography

DOI: 10.4328/ACAM.22028 Received: 2023-10-28 Accepted: 2023-12-04 Published Online: 2024-01-24 Printed: 2024-03-20 Ann Clin Anal Med 2024;15(Suppl 1):S47-53
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Introduction

Mandibular buccal bifurcation cyst (MBBC) is a rare inflammatory odontogenic cyst that was first described by Stoneman and Worth [1] in 1983 as 'mandibular infected buccal cyst'. In 1992 WHO classification [2], it was mentioned under the entity of paradental cyst as inflammatory collateral/mandibular infected buccal cyst.

MBBC has been classified under the inflammatory collateral cyst (ICC) in the 2017 [3] and the last 2022 WHO classifications (Available from: https://tumourclassification.iarc.who.int/chapters/52).

ICC comprises about 5% of all odontogenic cysts and includes paradental cysts and MBBCs (Available at: https://tumourclassification.iarc.who.int/chapters/52). MBBC comprise 35% of all ICC and is found on the buccal aspect of mandibular first or second molars and are often bilateral (up to 25%). Paradental cysts (60%) are found on the distobuccal aspect of mandibular third molars [4,5]. The etiology is inflammation in the pericoronal tissues. Cyst formation may be exacerbated by food impaction or by an enamel projection on the buccal aspect of the tooth [6,7].

MBBC is most commonly associated with the permanent mandibular first molar tooth, and secondly with mandibular second molar tooth [8]. The cyst can sometimes be seen bilaterally [9]. It is usually seen in children aged between 4-14 years, and the associated tooth has either recently erupted or partially erupted. Symptoms may increase as the size of the lesion progresses [10]. Sometimes it presents with very few symptoms, and sometimes pain, swelling, and suppuration can be observed [11]. The most common symptom is swelling in the buccal of the associated tooth. Its characteristic features are increased periodontal probing depth and buccal deviation of the crown of the associated tooth [12]. On clinical examination, the molar tooth may be missing, or the lingual cusps of the associated tooth may appear abnormally higher than the buccal cusps. The affected tooth is vital [9]. In the case of secondary infection, there may be pain, pus discharge, and facial asymmetry due to edema around the teeth [13,14].

In the radiographic findings of MBBC, there is a radiolucent lesion on the buccal surface of the affected tooth. The periodontal ligament space and lamina dura of the associated tooth is intact, and the root apices are displaced towards the mandibular lingual cortex. The lesion may cause expansion of the mandibular cortex and the formation of a periosteal reaction [15].

The histopathologic features of MBBC are not specific and consist of non-keratinized stratified squamous epithelium and inflamed connective tissue wall [10].

The general treatment option is the enucleation of the cyst without tooth extraction. Recurrence is not frequently observed [9]. Enucleation with tooth extraction and marsupialization can be other treatment options [16]. This article aims to present the clinical, radiological, and histological features and diagnostic criteria of MBBC and to add two new cases to the literature.

Material and Methods

Literature search was made on PubMed and Google Scholar. The following words were used for the search: (buccal bifurcation"OR"inflammatory collateral"OR paradental) AND cyst. The search was limited to articles in English. Case reports with clinical and radiographic features compatible with MBBC were included in the study. Case reports related to third molar teeth were excluded from the study because most showed paradental cyst features and others had not enough information.

Case Report

Case 1

A 9-year-old male patient was admitted to the clinic with the complaint of swelling. In the intraoral evaluation, it was observed that the bilateral mandibular first molars had partially erupted and there was swelling in the buccal gingiva. Radiolucent lesions associated with the right and left mandibular first molars were observed in the panoramic radiograph (Fig. 1). CBCT was taken to examine the effect of the lesion on the surrounding tissues. CBCT images showed well-circumscribed hypodense lesions starting from the furcation region of the bilateral mandibular first molars and extending to the inferior and anteroposterior directions. The expansion was observed in the buccal cortex. Periosteal reaction was not observed. Tipping was observed in the lingual direction in the roots of the associated teeth and in the buccal direction in the crowns (Figure 1). A preliminary diagnosis of bilateral mandibular buccal bifurcation cyst was made.

Case 2

An 8-year-old male patient was admitted to clinic due to pain and swelling. In the intraoral evaluation, it was observed that the right mandibular first molar tooth had partially erupted and there was swelling in the buccal gingiva. A radiolucent lesion associated with the right mandibular first molar tooth was observed in the panoramic radiograph (Fig. 2). CBCT was taken to examine the effect of the lesion on the surrounding tissues. On CBCT images, a well-circumscribed hypodense lesion was observed starting from the furcation region of the right mandibular first molars and extending to the inferior and anteroposterior direction. It was observed that the lesion caused expansion and destruction of the buccal cortex. Periosteal reaction was not observed. Lingual tipping was observed in the roots of the associated tooth (Figure 2). A preliminary diagnosis of mandibular buccal bifurcation cyst was made.

Both cases showed similar histopathologic features which were composed of the squamous non-keratinised epithelial lining in a characteristic arcading pattern and inflamed cyst walls (Figure 3). The patients were diagnosed with buccal bifurcation cyst with all clinicopathologic features. In the control panoramic radiograph of both cases, new bone formation areas were observed within the lesion, and there was no evidence of recurrence (Case 1: 8-month folllow-up, Case 2: 12-month follow-up).

Results

According to the results obtained in the literature search, a total of 81 MBBC cases were included in the study, and all findings were summarized in Table 1, Table 2, and Table 3. The age of the cases ranged from 6 to 17 years with the mean age of 8.4. Gender was specified in 75 cases and the majority of cases were male (n:49, 65.33%) while only 26 (34.66%) were female.

Symptom information was available in 52 cases. The most common symptom was swelling, reported in 27 (51.92%) cases. In 23 (44.23%) cases, both swelling and pain were present. Pain alone was observed in one patient, and pus discharge was observed in one patient. Vitality information was presented in 42 cases and 40 (95.23%) of them were vital. It was reported that one of the other 2 was devital and the other one had root canal treatment a month ago.

Location was reported in 80 cases. 27 (33.75%) cases were observed on the right side, 27 (33.75%) on the left side, and 26 (32.5%) bilaterally. In 80 cases, affected tooth number was given. The most common affected tooth was the mandibular first molar, which was reported in 69 (86.25%) cases, followed by the second molar tooth reported in 10 (12.5%) cases. In one case, the primary canine was associated with primary first and second molars. 23 bilateral cases were associated with the mandibular first molar (88.46%), while 3 cases were associated with the mandibular second molar (11.53%). Periosteal reaction was reported in 14 cases and no definite information was given in the remainder.

The treatment method was specified in 72 cases. Enucleation applied without tooth extraction was reported most frequently treatment method in 50 (69.44%) cases. The second most preferred method was enucleation with tooth extraction in 11 (15.27%) cases. While marsupialization was applied in 6 (8.33%) cases, the periodontal irrigation method was used in only one

Table 1. Cases of mandibular buccal bifurcation cyst from the literature.

Authors-Year	Patient sex/Age	Location in mandible/ Associated teeth	Vitality	Treatment/ Follow-up (month)	Recurrence	Symptom	Periosteal reaction
Current Study-2023	M/9	Bilateral/First molar	Vital	E/8	No	Swelling	No
	M/8	Right/First molar	Vital	E/12	No	Swelling and pain	No
Yaoran Liu [17]-2022	M/8	Left/First molar	NS	E/18	No	Swelling and pain	NK
	F/6	Left/First molar	NS	E/18	No	Swelling	NK
Ruddocks [18]-2022	M/11	Right/Second molar	NS	NK/NK	NS	NS	NK
	F/12	Bilateral/Second molar	NS	NK/NK	NS	NS	NK
	M/9	Bilateral/First molar	NS	E/NK	NS	NS	NK
	F/8	Left/First molar	NS	NK/NK	NS	NS	NK
	F/8	Right/First molar	NS	NK/NK	NS	NS	NK
	M/8	Right/First molar	NS	NK/NK	NS	NS	NK
	M/8	Left/First molar	NS	E/5	No	NS	NK
	F/9	Right/First molar	NS	NK/NK	NS	NS	NK
	M/9	Left/First molar	NS	NK/NK	NS	NS	NK
	M/8	Bilateral/First molar	NS	NK/NK	NS	NS	NK
Ashwag Aloyouny [19]-2021	M/7	Bilateral/First molar	Vital	TE and E/12	No	Swelling and pain	NK
dos Santos [16]-2021	F/6	Right/First molar	Vital	E/12	No	Swelling	NK
Dave [20]-2019	F/8	Right/First molar	Devital	TE and E/6	No	Swelling and pain	NK
	M/11	Bilateral/First molar	Vital	E/6	No	No	NK
	F/6	Bilateral/First molar	Vital	E/6	No	Swelling and pain(lift side)	+
Lima [13]-2019	M/7	Right/First molar	Vital	E/7	No	Swelling and pain	NK
Bautista [21]-2019	F/7	Bilateral/First molar	NS	E/NS		Swelling	NK
Kapoor [11]	F/13	Right/First molar	KT (1 month ago)	E/24	No	Pus drainage and	NK
						Localized swelling	
Dhanrajani [22]-2019	M/8	Left/Second molar	Vital	E/4	No	No	NK
Derindağ [23]-2018	M/10	Right/First molar	NS	E/NS	NS	Swelling and pain	+
Kim [24]-2018	M/8	Right/First molar	NS	E/24	No	Swelling and pain	NK
	M/9	Left/First molar	Vital	E/6	No	Swelling and pain	NK
Oenning [25]-2018	M/13	Left/Second molar	NS	E/NS	NS	No	NK
	F/17	Left/Second molar	NS	E/NS	NS	No	NK
Rana [26]-2018	F/6	Right/First molar	Vital	E/6	No	Swelling and pain	NK
De Grauve [27]-2018	F/8	Left/First molar	Vital	E/24	NS	No	+
	M/7	Right/First molar	Vital	TE and E/NS	NS	Swelling	+
	M/6	Left	Vital	E/NS	NS	Swelling and pain	+

F: Female, M: Male, NS: Not stated, E: Enucleation, NK: Not known, TE: Tooth extraction, NP: No procedure, MA: Marsupialization.

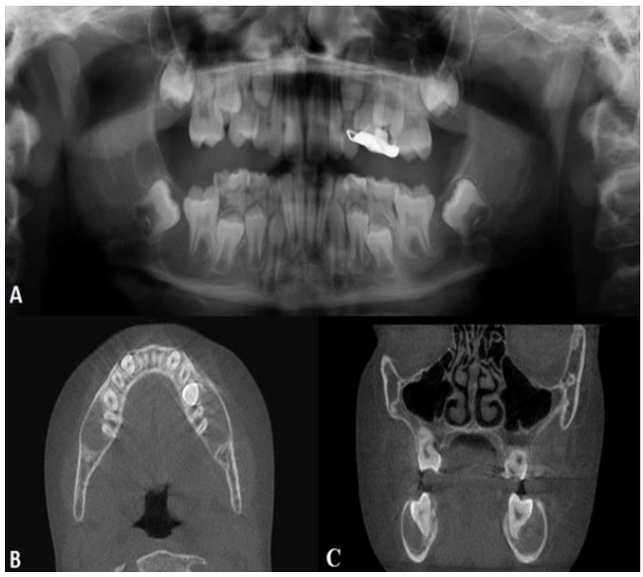

Figure 1. Radiographic Images of Case 1: (A) Panoramic radiography, (B) CBCT axial section, (C) CBCT coronal section.

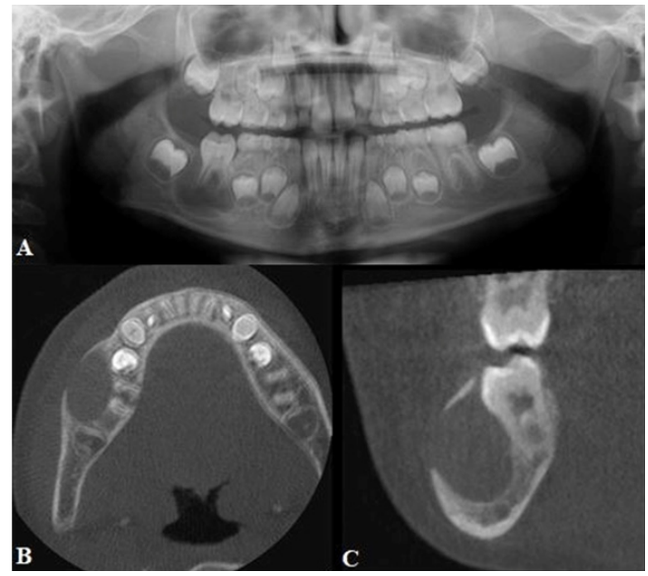

Figure 2. Radiographic Images of Case 2: (A) Panoramic radiography, (B) CBCT axial section, (C) CBCT coronal section.

Table 2. Cases of mandibular buccal bifurcation cyst from the literature (continued).

Authors-Year	Patient sex/Age	Location in mandible/ Associated teeth	Vitality	Treatment/ Follow-up (month)	Recurrence	Symptom	Periosteal reaction
Omami and Al Yafi [28]-2016	M/15	Right/Second molar	NS	NS/NS	NS	Swelling and pain	+
Levarek [29]-2014	M/7	Left/First molar	Vital	E/24	No	Swelling	NK
	F/6	Left/First molar	NS	E/9	No	Swelling	+
	F/7	Left/First molar	NS	E/5	No	Swelling and pain	NK
Friedrich [30]-2014	M/6	Left/First molar	Vital	E/15	NS	Swelling	NK
Borgonovo[31]-2014	M/6	Right/First molar	-	E/NS	NS	Swelling	NK
Islerr [32]-2013	F/8	Bilateral/First molar	Vital	E/18	No	No	NK
	M/9	Right/First molar	Vital	E/2	No	Swelling and pain	NK
Borgonovo[33]-2013	F/14	Left/Second molar	Vital	E/6	NS	No	NK
Ramos [10]-2012	M/9	Bilateral/First molar	Vital	E/12	No	Swelling and pain	NK
Boffano [34]-2012	NS/9	Bilateral/First molar	Vital	E/6	No	Swelling	NK
Borgonovo[35]-2012	M/8	Bilateral/First molar	Vital	E/12	No	Swelling	NK
Corona-Rodriguez [36]-2011	M/7	Bilateral/First molar	Vital	E (right side) NP left side)/6	No	Swelling	NK
Zadik [37]-2011	NS/7	Bilateral/First molar	Vital	NS	NS	Swelling and pain	NK
Santos [38]-2011	NS/8	Left/First molar	Vital	E/6	No	Swelling	NK
Lizio [39]-2011	F/8	Bilateral/First molar	Vital	MP/48	No	No	NK
	M/7	Right/First molar	Vital	MP/24	No	No	NK
	F/10	Left/First molar	Vital	MP/60	No	No	NK
	M/8	Left/First molar	Vital	MP/24	No	No	NK
	F/10	Left/First molar	Vital	MP/12	No	No	NK
Chrcanovic[40]-2011	F/6	Right/First molar	Vital	E/16	No	Swelling	+
Borgonovo[41]-2010	M/7	Left/First molar	NS	E/12	No	Swelling	NK
	M/8	Left/First molar	Vital	E/12	No	Swelling and pain	NK
Thikkurissy [42]-2010	M/7	Right/First molar	Vital	E/14	No	Swelling and pain	+
Iatrou [43]-2009	M/7	NS/NS	NS	E/NS	NS	NS	NK
	M/9	Left/First molar	NS	TE and E/NS	NS	NS	NK
	M/8	Right/First molar	NS	TE and E/NS	NS	NS	NK
	M/8	Right/First molar	NS	TE and E/NS	NS	NS	NK

F: Female, M: Male, NS: Not stated, E: Enucleation, NK: Not known, TE: Tooth extraction, NP: No procedure, MA: Marsupialization.

case. It was observed that 4 (5.55%) cases with small sizes were followed up without any procedure. No recurrence was reported in control radiographs in 47 cases. Overall, there were no recurrences reported in any of the cases in the literature.

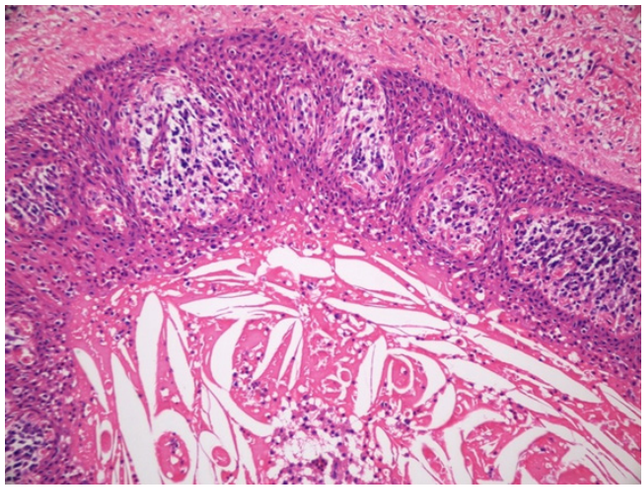

Figure 3. An inflamed fibrous wall lined by non-keratinized squamous epithelium with arcading rete hyperplasia and has no specific features. Please note the cholesterol deposition in the lumen (H&E x200).

Discussion

The clinical-radiographic differential diagnosis of MBBC includes lateral periapical cyst, lateral periodontal cyst, dental follicle, dentigerous cyst, periodontal abscess, odontogenic keratocyst, and eosinophilic granuloma [13]. Periodontal abscess and Langerhans cell histiocytosis can also cause an inflammatory periosteal reaction like MBBC. The most characteristic feature of MBBC in differentiating it from other lesions is that it pushes the roots of the affected tooth into the lingual. A dentigerous cyst is also in the differential diagnosis. However, the epicenter of a dentigerous cyst is different because a MBBC starts near the bifurcation region of the tooth and does not surround the crown, unlike a dentigerous cyst [9]. Hyperplastic dental follicles are enlarged normal follicles, surrounding the crown of the developing tooth and following the general outline of that tooth crown. Odontogenic keratocyst may be pericoronal and may resemble a dentigerous cyst in the early stages. It may appear similar to MBBC if adjacent to the roots but causes little enlargement of the mandible. A lateral radicular cyst is an odontogenic inflammatory cyst associated with a nonvital tooth. The epicenter is located close to the apex. Large cysts may extend into the furcation area and may be confused with MBBC. Large cysts may extend into the furcation area and may be confused with MBBC. However, MBBC is more expansile than a radicular cyst of the same size. A lateral periodontal cyst is an odontogenic cyst that occurs lateral to the tooth roots. It

Table 3. Cases of mandibular buccal bifurcation cyst from the literature (continued).

Authors-Year	Patient sex/Age	Location in mandible/ Associated teeth	Vitality	Treatment/ Follow-up (month)	Recurrence	Symptom	Periosteal reaction
Gallego [44]-2007	M/8	Bilateral/First molar	Vital	E (left side) NP (right side)/12	No	Swelling and pain	NK
Shohat [45]-2003	F/11	Right/Second molar	Vital	TE and E/>24	No	Swelling	NK
	F/8	Right/First molar	Vital	TE and E/>24	No	Swelling and pain	NK
	F/8	Right/First molar	Vital	E/36	No	Swelling	NK
	M/13	Bilateral/First molar	Vital	E/14	No	Swelling	NK
	M/8	Bilateral/First molar	Vital	E/>24	No	Swelling	NK
David [46]-1998	M/8	Bilateral/First molar	NS	NP/15	NS	Swelling (Bilateral)	NK
	M/9	Bilateral/First molar	NS	NS (right side);NP (left side)/9	NS	No	NK
	M/7	Bilateral/First molar	NS	Periodontal irrigation /NS	NS	Swelling and pain	NK
Martinez-Conde [47]-1995	M/11	Bilateral/Second molar	NS	TE and E /NS	NS	Swelling (Bilateral)	NK
Bohay [48]-1992	M/7	Bilateral/First molar	-	E/ 8	-	-	NK
Packota [49]-1990	M/6	Bilateral (asynchronous) / First molar	NS	E/ 6 (right side); notstated (left side)	No	Swelling (Bilateral)	+
	NS/7	Left/First molar	NS	E/ NS	NS	Toothache and swelling	+
	NS/7	Right/First molar	NS	E/ NS	NS	Swelling	+
	NS/8	Right/First molar	NS	E/ NS	NS	Swelling and pain	+
	M/8	Left/First molar	NS	E/ 9	NS	Pain	NK
Camarda [50]-1989	M/9	Right/ Primary canine, and primary first and second molars	Vital	TE and E/ 60	No	Swelling	NK
	M/9	Left/First molar	NS	E/ NS	NS	Swelling	NK
Trask [51]-1985	F/6	Left/First molar	Vital	TE and E/ 9	NK	NK	+
Swerdloff [52]-1980	F/7	Bilateral/First molar	NK	E/ 6	NK	NK	NK
Stanback [53]-1970	M/9	Bilateral/First molar	NS	MA/24	No	Swelling (Bilateral)	NK

F: Female, M: Male, NS: Not stated, E: Enucleation, NK: Not known, TE: Tooth extraction, NP: No procedure, MA: Marsupialization.

usually appears as inter-radicular radiolucency. The mandibular premolar region is the most common site. It is seen in the older age groups [15].

When MBBC is visualized on conventional radiographs, it presents as a normal lamina dura and superposed radiolucency on tooth roots. Since the lesion is non-endodontic origin, the periodontal space surrounding the roots and the lamina dura is observed as intact [13]. On periapical and panoramic radiographs, the cyst may sometimes appear to be located slightly distal to the furcation of the associated tooth [9]. However, it is not always easy to diagnose with conventional radiographs [42]. With cone-beam computed tomography (CBCT), the borders of the lesion, its effects on the surrounding tissues, and its characteristic features can be displayed more clearly. It is often observed as unilocular, well-defined radiolucency, starting from the furcation area of the associated tooth and spreading to the apex. Pompura et al. reported that cortical periosteal reaction was observed in 68.8% of MBBC cases [12,54]. CBCT may also be successful in demonstrating buccolingual enlargement caused by the cyst and lingual deviation of the buccal root of the crown of the associated tooth. In addition, occlusal radiographs can be used in MBBC imaging [12,16]. The lingual tipping of the roots, cortex expansion, and periosteal reaction can be observed on the occlusal radiography [9].

MBBC histopathology is indistinguishable from radicular cyst. Because of its histopathologic similarity with inflammatory odontogenic cysts, the diagnosis should be made by clinical and radiological evaluation [16].

There are different approaches to the treatment method. There are two main approaches for the treatment of MBBC in the literature. The first is enucleation and curettage without tooth extraction, and the second is tooth extraction with curettage and enucleation [16]. David et al. [46] suggested a more conservative method described as 'micro-marsupialization'. In this method, periodontal probing and irrigation of the buccal pocket with saline or hydrogen peroxide are performed. Daily saline irrigation is then continued. It was thought that creating a small opening with periodontal probing would reduce the pressure of the cyst and allow it to regress spontaneously. David et al. [46] also mentioned another conservative method called 'automarsupialization' and 'self-dissolution'. This method is based on follow-up and states that microtraumas can cause spontaneous remission of the lesion. High bone remodeling capacity and rapid healing in children of the relevant age group may explain this method [16]. Zadik et al. [37] and Corona-Rodriguez et al. [36] mentioned spontaneous resolution cases in their studies. In small and asymptomatic cases, marsupialization may be a treatment option for the pediatric age group, thanks to its rapid bone remodeling capacity. However, larger lesions require surgery [55]. Some authors suggest that pulp vitality is at risk when tooth formation is complete, and endodontic treatment or extraction of the tooth is indicated when necessary. The most accepted approach in the treatment of MBBC is enucleation and curettage without tooth extraction. This conservative surgical method aims to protect the first and second molar teeth, which are important to keep in the mouth [13]. When the lesion is treated appropriately, it rarely recurs and has a good prognosis [56].

Conclusions

In this paper, we reported two additional cases consistent with most of the literature in terms of gender (male), common symptoms (swelling and pain), affected tooth (mandibular permanent first molar tooth) and prognosis (no recurrence). In addition, the clinical signs, symptoms, and treatment features of MBBC cases reported in the literature were complied. Knowing this entity, especially in childhood, will guide clinicians in protecting the affected tooth while performing cyst treatment in most of the cases.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and Human Rights Statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or compareable ethical standards.

Funding: *None*

Conflict of Interest

The authors declare that there is no conflict of interest.

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How to cite this article:
Rabia Duman Tepe, Hulya Cakir Karabas, Merva Soluk Tekkesin. *Mandibular Buccal Bifurcation Cyst with Radiologic and Histopathologic Features: Two Case Reports and a Literature Review. Ann Clin Anal Med 2024;15(Suppl 1):S47-53*